Case Report

Acute Catatonia With Urinary Incontinence in an Adolescent: A Case Report and Review of the Literature

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ABSTRACT

Background: Few studies have investigated catatonia or adequately elucidated its epidemiology or pathophysiology. Catatonia is a debilitating neuropsychiatric syndrome.

Case presentation: In this case report, we present a 15-year-old boy who was brought to the emergency room with weakness, gait disturbance, and urinary incontinence.

Conclusion: The findings of this investigation indicate that in cases where medicinal intervention has proven ineffective, electroconvulsive therapy (ECT) seems to be a commendable substitute form of treatment.

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Introduction

Catatonia is a potentially life-threatening psychomotor dysregulation syndrome where patients cannot move normally even if they have the physical capacity. Unfortunately, catatonia could be underdiagnosed and undertreated in the pediatric population [1]. The prevalence of pediatric catatonia in clinical psychiatry ranges from 0.6% to 17% [2]. To confirm the diagnosis of catatonia, the presence of 3 out of 12 cardinal symptoms is necessary based on the diagnostic and statistical manual of mental disorders, fifth edition (DSM-5): Mutism (verbal unresponsiveness), waxy flexibility (allowing the limbs to be shaped at the examiner's will), echolalia (repetition of the sounds and words produced by others), echopraxia (imitation of physical gestures), negativism (involuntary resisting any efforts), catalepsy (maintaining peculiar postures for a long period), stupor (unresponsiveness to stimulation), agitation (a state of nervous excitement), stereotypies (repetitive motor activity), mannerisms (exaggerated motor behavior), grimacing, and posturing [1].

The mechanisms underlying catatonia are not currently well understood. Dysfunctions of brain circuits between areas such as the orbitofrontal cortex, prefrontal cortex, supplementary motor area, thalamus, and cerebellum could explain motor symptoms of catatonia [3]. Reduced activity of gamma-aminobutyric acid type A and N-methyl-d-aspartate receptors as the origin of the motor and affective symptoms has also been suggested. Dopamine and other neurotransmitter dysregulation, abnormal neuronal activity, and dysfunction of oligodendrocytes have also been suggested in the pathophysiology of catatonia [3]. We report an adolescent with acute catatonia and urinary incontinence.

Case Presentation

In this case report, we present a 15-year-old boy with catatonia symptoms. The source of history was the patient's biological father and stepmother. They both seemed reliable. The patient and his parents signed a written consent form before reporting the patient's symptoms. The patient was a 9th-grade student working in a mechanic repair shop. He was brought to the Emergency Department with weakness, gait disturbance, and urinary incontinence. Gradually, stupor, mutism, and insomnia emerged. He could not walk or speak. He had no history of any substance or drug use. Intravenous diazepam was partially effective in the remission of some of his symptoms. The day after the diazepam injection, his symptoms relapsed. He was taken to another Emergency Department the second time and received only oral melatonin. The symptoms became exacerbated, and he refused to eat or drink anything. He was staring at the water and was not washing himself in the bathroom. He was brought to the Emergency Department for the third time and finally admitted to the Pediatric Neurologic Ward. Although the neurological examination and brain imaging were normal, he still could not walk or speak. Other signs and symptoms were urinary incontinence, crying, and depressed mood. The patient was discharged by her parents against medical advice. The symptoms continued, and the parents had to take the patient to a psychiatric hospital.

The patient had normal development without a history of psychiatric problems. He had no history of enuresis, elevated mood, irritability, hyper-sexuality, or physical or verbal aggression. In the last month before admission, he complained of boredom, forgetfulness, and poor concentration, but he still went to school and did his homework. He got good grades and had no behavioral disturbances or conduct problems at school. The patient had no history of any medical conditions like head trauma, seizure, kidney, liver, or thyroid disease, etc. Drug and substance history was negative.

His biological mother had died of cancer when the patient was 9. Father had no history of psychiatric disorder. The patient's uncle had a diagnosis of intellectual disability and a history of psychiatric admission. Two of his cousins had a history of schizophrenia.

He had no history of being abused. Although he was a shy boy with difficulty in making friendships, he never met the criteria for autism spectrum disorder.

Mental state exam

On presentation, the patient could enter the examination room with the help of two other persons. He appeared depressed, agitated, and uncooperative. During the interview, he was mute and entirely unresponsive to the examiner's voice. Sometimes, he whispered something with a low tone of speech. He repeated, "they bothered me." The content of his thoughts could not be adequately evaluated. We did not know if he had any suicidal ideation, obsession, or delusion, but it appeared that he had paranoid ideation. He had frequent response latency and seemed to have thought of blocking. Assessment of his perception was very difficult. He showed severe psychomotor retardation and maintained a peculiar and unusual posture for hours. He al-

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Table 1. Patient's physical examination

Blood Pressure	Temperature	Pulse Rate	Weight	Height	Pupil
125/80 mm Hg	36.5°C	88	51 kg	167 cm	Mydriasis, reactive to light

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lowed his limbs to be shaped at the examiner's will. The patient's presentation was not fixed and changed from one visit to another. He did not react to something happening and sometimes actively resisted instructions. There was no tattooing or scars on his exposed skin. His insight was 1/6. Table 1 and 2 show the physical exams and laboratory results.

Serum creatine phosphorous kinase (CPK) was in the normal range. Urine analysis (U/A) was normal, and urine culture was negative. Electroencephalography (ECG) showed normal sinus rhythm. Viral studies and N-methyl-d-aspartate (NMDA) receptor antibodies were negative. Lumbar puncture and chest x-ray revealed no abnormality. EEG, brain magnetic resonance imaging (MRI), and brain CT had no abnormal findings. Lab data (at physical, psychiatric admission). Urine screening for drug and substance was negative except for benzodiazepine. Abdominal and pelvic ultrasound was normal. His medical workups revealed no genitourinary infection or anomaly.

Lorazepam was started according to the catatonic symptoms, but he was distrustful of the staff and refused to eat food or take medication. The parents gave written informed consent to provide the patient with electroconvulsive therapy (by ECT instrument, Thermatron® System IV Manufacturer: Lake Bluff, IL, USA). Standard bilateral electroconvulsive therapy (ECT) was administered on temporal regions. After receiving the first ECT session, the patient's compliance was improved. The patient received lorazepam 6 mg per day, but the catatonic symptoms continued. Finally, he received 6 bilateral standard ECT sessions every other day. During the treatment sessions, the patient's mood gradually increased. The catatonic symptoms, as well as urinary incontinence, were completely resolved. Third week after the hospitalization, the patient showed elevated mood, unexplained laughing, talkativeness, inflated self-esteem, increased energy, and insomnia despite receiving lorazepam. He was started on lithium carbonate 300 mg three times a day. Therapeutic adherence was improved, and the symptoms resolved gradually.

Finally, the diagnosis of bipolar disorder with catatonic features was made, and the patient was discharged with lorazepam 6 mg/daily and lithium 300 mg three times daily. The patient was revisited one week after discharge and one month and two months later for follow-up. Medication adherence was good. Symptoms of catatonia and urinary incontinence did not recur. He was euthymic, returned to the previous psychosocial function, and was regularly going to school.

Table 2. Patient's laboratory data

Biochemical Assay	1st Admission	2nd Admission
WBC (4.5-11.0x10^9/L)	13500	4500
PMN	74%	61%
Lymphocyte	16%	36%
ESR (mm/hr)	50	8
CRP (mg/L)	26	Negative
AST (U/L)	25	23
ALT (U/L)	12	13
ALKP (U/L)	376	368
TSH (mIU/L)	3.5	3.5

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Abbreviations: PMN: Polymorphonuclear leukocyte; ESR: Erythrocyte sedimentation rate; CRP: C-reactive protein; AST: Aspartate aminotransferase; ALT: Alanine transaminase; ALKP: Alkaline phosphatase; TSH: Thyroid stimulating hormone.

Discussion

Catatonia is widely regarded as a rare neuropsychiatric syndrome that affects the pediatric and adolescent populations [1]. Typically, its onset is acute, making it easily confused with delirium, epilepsy, and other acute neurological disorders [2]. The most common medical conditions underlying catatonia are infections (e.g. viral encephalitis, typhoid fever, and toxoplasmosis), non-convulsive status epilepticus, auto-immune diseases (e.g. systemic lupus erythematosus and anti-NMDA receptor encephalitis), drug-induced states, and metabolic disorders [4, 5]. Although the cardinal symptoms of catatonia seem to be similar across all ages, some symptoms may be more common in pediatric patients, including social withdrawal, refusing to eat or drink, repetitive movements, and regressive symptoms, such as urinary incontinence [2]. An interesting aspect of the current case is that our patient presented with urinary incontinence at the onset of catatonia. Furthermore, the incontinence was resolved when catatonic symptoms improved with appropriate treatment. The reported case study is intriguing for several reasons. There are only a few cases of catatonia presenting with urinary incontinence [6]. The underlying pathophysiology of urinary incontinence associated with catatonia is not well-known. Also, according to DSM-5 criteria, urinary incontinence is not yet considered a symptom of catatonia [6].

However, the number of studies addressing such issues is minimal: One such study, conducted in 2021, reported catatonia in a 12-year-old girl with post-traumatic stress disorder [7]. Benzodiazepines, especially lorazepam, could stimulate gamma-aminobutyric acid activity. They are considered the first-line treatment for improving catatonic symptoms [2].

Schizophrenia and mood disorders are the most common psychiatric disorders associated with catatonia in children and adolescents. Mood disorders' symptoms may present after the improvement of catatonia [1, 8, 9]. Neurodevelopmental disorders like autism spectrum disorder, Down syndrome, and Tourette syndrome are also associated with higher rates of catatonia [10]. In our case, psychiatric symptoms were unmasked after partial remission of catatonic symptoms. He presented with elevated mood, increased tone and rate of speech, increased energy, inflated self-esteem, and decreased need to sleep. The final diagnosis of bipolar mood disorder with catatonic features was made.

Our patient had a normal growth without any psychiatric or neurodevelopmental disorder. All his medical workups were within the normal range, and none showed an abnormality. His neurological examination and EEG were also in the normal range. Our patient had poor compliance with pharmacotherapy. Although he received lorazepam 6 mg daily, only some of his catatonic symptoms were partially remitted. In patients with resistance to lorazepam, a thorough medical evaluation is essential to uncover the underlying cause [1, 9, 11].

After receiving the first ECT session, the patient's adherence to treatment was improved. Finally, he received 6 bilateral standard ECT sessions every other day. Lorazepam was administered thrice daily, with the last daily dose prescribed 12 hours before each ECT session.

Electroconvulsive therapy is an effective intervention in most subtypes of catatonia [1, 2, 9]. Some studies report a synergistic effect of treatment with both lorazepam and ECT. Electroconvulsive therapy can lower cortical excitability and potentiate the effect of lorazepam [11]. Therefore, recognizing catatonia in its early stages and starting effective therapeutic interventions using a multi-disciplinary approach is essential and can reduce the risk of complications like dehydration, malnutrition, pressure ulcers, infections, aspiration pneumonia, pulmonary embolism, and deep vein thrombosis [1, 9]. Consequently, early treatment of catatonia can reduce patients' risk of complications.

Conclusion

In conclusion, this is a case report of acute catatonia with urinary incontinence in an adolescent and reminds us that patients with catatonia may present with unusual clinical manifestations. The results of this study suggest that when drug therapy has failed, ECT appears to be a good alternative treatment. We recognize the limitations of this case report and acknowledge that future studies should focus on the pathophysiology of pediatric catatonia and the best therapeutic interventions.

Ethical Considerations

Compliance with ethical guidelines

All ethical principles are considered in this article.The patient has duly furnished written informed consent to disseminate this manuscript and to conduct material sampling.

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Authors contributions

All authors equally contributed to preparing this article.

Conflicts of interest

The authors have no other notable affiliations or financial involvements with any organization or entity that might have a financial interest in or may cause an economic conflict with the subject matter or materials discussed in the manuscript, apart from those that have been mentioned.

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